






# Physical or Occupational Therapy Use in Systemic Sclerosis: A Scleroderma Patient-centered Intervention Network Cohort Study

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**ABSTRACT. Objective.** Systemic sclerosis (SSc) is characterized by significant disability because of musculoskeletal involvement. Physical and occupational therapy (PT/OT) have been suggested to improve function. However, the rate of PT/OT use has been shown to be low in SSc. We aimed to identify demographic, medical, and psychological variables associated with PT/OT use in SSc.

**Methods.** Participants were patients with SSc enrolled in the Scleroderma Patient-centered Intervention Network (SPIN) Cohort. We determined the rate and indication of PT/OT use in the 3 months prior to enrollment. Multivariable logistic regression was used to identify variables independently associated with PT/OT use.

**Results.** Of the 1627 patients with SSc included in the analysis, 23% used PT/OT in the preceding 3 months. PT/OT use was independently associated with higher education (OR 1.08, 95% CI 1.04–1.12), having moderately severe small joint contractures (OR 2.09, 95% CI 1.45–3.03), severe large joint contractures (OR 2.33, 95% CI 1.14–4.74), fewer digital ulcerations (OR 0.70, 95% CI 0.51–0.95), and higher disability (OR 1.54, 95% CI 1.18–2.02) and pain scores (OR 1.04, 95% CI 1.02–1.06). The highest rate of PT/OT use was reported in France (43%) and the lowest, in the United States (17%).

**Conclusion.** Despite the potential of PT/OT interventions to improve function, < 1 in 4 patients with SSc enrolled in a large international cohort used PT/OT services in the last 3 months. Patients who used PT/OT had more severe musculoskeletal manifestations and higher pain and disability. (First Release August 15 2019; J Rheumatol 2019;46:1605–13; doi:10.3899/jrheum.181130)

## Key Indexing Terms:

SYSTEMIC SCLEROSIS  
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Systemic sclerosis (SSc; also called scleroderma) is a chronic systemic disease characterized by dysregulated fibrosis, autoimmunity, inflammation, and vasculopathy<sup>1</sup>. Musculoskeletal involvement is nearly universal in SSc. Skin fibrosis, joint and muscle pain, arthritis, hand deformities, joint

contractures, and reduced range of motion are common manifestations and result in significant disability<sup>2</sup>. Hand pain and joint stiffness are among the 5 highest-rated symptoms and are described by more than 80% of patients with SSc<sup>3</sup>. Skin fibrosis of the face and oral tissues causes difficulties with eating, speaking, dental care, and oral hygiene<sup>4</sup>.

Musculoskeletal rehabilitation and physical and occupational therapy (PT/OT) are recommended for the management of musculoskeletal impairment in SSc<sup>5</sup>. Multiple rehabilitation techniques including range of motion exercises, connective tissue massages, joint manipulation, splinting, heat/paraffin wax baths, and generalized PT have been suggested to reduce pain and improve joint motion in small randomized control trials, case reports, and case series<sup>5</sup>. Similarly, hand and orofacial exercises have been suggested to improve gingival health<sup>6,7</sup>. A trial of 220 patients with SSc found that a 4-week PT program significantly reduced disability 1 month post-randomization, although there was not an effect on disability at the 12-month followup<sup>8</sup>.

Variable rates of PT/OT use have been reported in the literature with most studies showing < 50% of patients with SSc using PT/OT despite it being one of the primary available interventions to address musculoskeletal manifestations<sup>9,10,11</sup>.

In our study, we aimed to determine the rates and indications of PT/OT use among patients with SSc enrolled in one of the largest SSc cohorts worldwide, the Scleroderma Patient-centered Intervention Network (SPIN) Cohort. We also aimed to identify demographic, medical, and psychological variables associated with the use of these services.

## MATERIALS AND METHODS

*Patients and procedures.* Our present study included a convenience sample of patients enrolled in the SPIN Cohort. As previously described<sup>12</sup>, this large international cohort is composed of patients recruited from more than 40 centers in Canada, the United States, and Europe. To be eligible for enrollment, patients must have been classified as having SSc according to the 2013 American College of Rheumatology/European League Against Rheumatism classification criteria<sup>13</sup>, be 18 years of age or older and fluent in English, Spanish, or French, and have the ability to provide informed consent and respond to SPIN questionnaires online. Medical variables are completed by the SPIN physician or coordinator, initiating enrollment in the SPIN Cohort. Patients are then invited by e-mail to register and complete the SPIN Cohort questionnaires online. Included in our study were SPIN patients who completed baseline questionnaires from January 2014 through September 2017. The SPIN Cohort study was approved by the institutional review boards (IRB) of the Jewish General Hospital, Montreal, Quebec, Canada (ethics protocol # CODIM-FLP-12-123), the Hospital for Special Surgery, New York, USA (IRB study # 2014-326), and all other participating centers. All patients provided informed consent.

*PT/OT use.* Patients were asked to answer 3 questions on PT/OT use:

1. "In the last 3 months, have you seen a physical therapist, physiotherapist, occupational therapist, ergotherapist, kinesiotherapist or other health care professional for rehabilitation services?"
2. "If yes, what was this for?" Patients could pick one or more options from hands, feet, mouth/face, wound care, activities of daily living, other.
3. "If yes, how many times in the last 3 months did you receive rehabilitation services?"

*Demographic variables.* Demographic variables were completed by patients and included race/ethnicity, years of education, current occupation, and housing location. Race/ethnicity were entered differently in different countries and were categorized according to the corresponding country's definitions. A consolidated race/ethnicity variable that included white, black, and other was created for statistical analysis. Enrolling physicians identified the site of enrollment, sex, and date of birth at baseline.

*Medical variables.* Disease-specific variables included disease subtype (diffuse or limited), duration since first non-Raynaud disease manifestation, the presence of Raynaud phenomenon, modified Rodnan skin score (mRSS), digital ulcerations (DU), tendon friction rubs, joint contractures, cardiopulmonary disease, and overlap syndromes. Diffuse SSc was defined as skin sclerosis involving the limbs proximal to the elbows and knees and/or the chest and/or trunk at any time, whereas limited SSc was defined as SSc confined to the limbs distal to the elbows. The mRSS is a clinical measure of skin thickness from 0 to 51, with higher scores indicating more severe thickness. The presence of overlap syndromes with rheumatoid arthritis (RA), systemic lupus erythematosus, and/or idiopathic inflammatory myositis was also determined.

*Disability measures.* Patients completed the Scleroderma Health Assessment Questionnaire (SHAQ) and the Cochin Hand Function Scale (CHFS-II). The SHAQ assesses physical disability with a composite score from 0 to 3, with higher scores indicating greater disability<sup>14,15</sup>. The CHFS-II assesses hand disability with a score from 0 to 90, with higher scores indicating higher disability<sup>15,16</sup>. The SHAQ and CHFS-II have been validated in patients with SSc<sup>14,15,16</sup>.

*Psychological measures.* To assess symptoms of depression, patients completed the Patient Health Questionnaire (PHQ-8). Scores range from 0 to 24, with higher scores indicating more depressive symptoms<sup>17</sup>. Patients also completed the Patient Reported Outcomes Measurement Information System (PROMIS-29) measure version 2, which assessed 7 patient-reported outcome domains including the anxiety, fatigue, and pain intensity domains used in our study. Scores are standardized with a mean of 50 and an SD of 10, with the mean score representing the average of a general US population and higher scores reflecting more of the measured domain<sup>18</sup>. The Satisfaction with Appearance (SWAP) scale was used to measure body image distress. Scores can range from 0 to 84, with higher scores indicating greater dissatisfaction with appearance<sup>19</sup>. The Social Interaction Anxiety Scale-6 was used to assess distress due to social interactions by rating patients' experience in social situations from 0 to 24, with higher scores indicating higher social anxiety symptoms<sup>20</sup>. Patients also completed the Self-Efficacy for Managing Chronic Disease (SEMCD) scale to assess their confidence in self-managing disease-specific symptoms. Scores ranged from 1 to 10, with higher scores indicating higher self-efficacy. These scores have been validated in SSc<sup>18,19,21,22,23,24,25</sup>.

*Statistical analysis.* Descriptive analyses included means, SD, minimum and maximum for continuous variables, and frequencies and percentages for discrete variables. Prior to initiation of inferential analyses, data completeness and normality for continuous measures was evaluated. Demographic, psychological, disability, and medical variables were compared between patients who participated in PT/OT in the prior 3 months and those who did not using a chi-square test and independent samples t test. Multivariable binary logistic regression was subsequently used to identify variables independently associated with PT/OT use through an *a priori* defined model that included age, sex, education level, employment status, disease subset, disease duration, joint contractures, DU, SHAQ, the PROMIS domain on pain intensity, PHQ-8, and SEMCD. Country of enrollment was analyzed after predefined variables were identified. To improve the precision of measurement estimates and fit of the data in the model, backward stepwise procedure was used to build best-fitting parsimonious models to best identify predictors of PT/OT usage. Missing variables were not imputed or replaced. P values of 0.05 or below were considered statistically significant. All statistical analyses were performed using SAS version 9.3.

## RESULTS

**Sample characteristics.** At the time of data extraction, 1641 patients had completed baseline assessment, of which 1627 patients answered the PT/OT use questions and were included in the analysis. Of the patients, 43% were from the United States, 27% from Canada, 17% from France, 10% from the United Kingdom, and 2% from Spain. Patients had a mean ( $\pm$  SD) age of  $54.9 \pm 12.5$  years. Female patients constituted 87% of the patients and diffuse SSc was reported in 41% of patients. Mean disease duration was  $11.4 \pm 8.8$  years. The mean mRSS was  $7.8 \pm 8.4$ . Interstitial lung disease and pulmonary arterial hypertension were reported in 36% and 10% of patients, respectively.

**PT/OT use.** Of the 1627 patients, 381 (23%) used PT/OT in the 3 months prior to enrollment in SPIN. The mean number of times these services were used among patients who used it in the last 3 months was  $9.8 \pm 10.7$ . The rate of PT/OT use varied among the different countries and was highest in France (43%), followed by Spain (28%), Canada (23%), United Kingdom (19%), and United States (17%). In the entire cohort, hand PT/OT was the most common indication and was reported by 59% of patients. Feet and activities of daily living were other common indications and were reported by 27% and 30% of patients, respectively (Figure 1).

**Factors associated with PT/OT use.** In bivariate analyses, no differences were observed in age, sex, race, education, or housing location among patients who used PT/OT in the 3 months prior to enrollment (Table 1). Compared to employed patients, we observed a higher rate of unemployed (28% vs 20%,  $p = 0.01$ ) and disabled (36% vs 20%,  $p < 0.01$ ) patients in the PT/OT use group. While all other sites reported a higher percentage of PT/OT use compared to the United

States, only Canada (23% vs 17%,  $p = 0.02$ ) and France (43% vs 17%,  $p < 0.01$ ) had statistically significant higher use of PT/OT services.

Patients who used PT/OT were more likely to have the diffuse compared to the limited form of the disease (29% vs 19%,  $p < 0.01$ ) and early ( $\leq 3$  yrs) compared to late ( $> 3$  yrs) disease (29% vs 23%,  $p = 0.04$ ). Compared to patients who did not use these services, they were also more likely to have shorter disease duration ( $10.4 \pm 8.6$  yrs vs  $11.7 \pm 8.8$  yrs,  $p = 0.02$ ) and higher mRSS ( $10.0 \pm 9.9$  vs  $7.1 \pm 7.8$ ,  $p < 0.01$ ). The PT/OT group had more tendon friction rubs in the past (27% vs 21%,  $p < 0.01$ ) and currently (31% vs 21%,  $p = 0.049$ ) compared to never, moderately severe small joint contractures (37% vs 19%,  $p < 0.01$ ) and severe small joint contractures (42% vs 19%,  $p < 0.01$ ) compared to no/mild small joint contractures, and moderately severe large joint contractures (40% vs 21%,  $p < 0.01$ ) and severe large joint contractures (37% vs 21%,  $p = 0.01$ ) compared to no/mild large joint contractures. Patients who used PT/OT were slightly more likely to have interstitial lung disease than not (27% vs 22%,  $p = 0.02$ ). No differences in Raynaud phenomenon, DU, pulmonary arterial hypertension, and overlap syndromes were seen.

Regarding functional impairment (Table 2), patients who used PT/OT had higher hand dysfunction (CHFS-II  $20.3 \pm 18.9$  vs  $11.6 \pm 14.6$ ,  $p < 0.01$ ) and functional disability scores (SHAQ  $1.0 \pm 0.7$  vs  $0.7 \pm 0.7$ ,  $p < 0.01$ ). These patients were also found to have statistically significantly more pain ( $59.3 \pm 8.6$  vs  $54.5 \pm 9.7$ ,  $p < 0.01$ ), fatigue ( $57.5 \pm 10.1$  vs  $54.4 \pm 11.2$ ,  $p < 0.01$ ), and anxiety ( $54.1 \pm 10.2$  vs  $51.3 \pm 10.0$ ,  $p < 0.01$ ) as measured by PROMIS-29; higher depression symptoms (PHQ-8  $7.7 \pm 5.8$  vs  $5.9 \pm 5.3$ ,  $p < 0.01$ ); and more

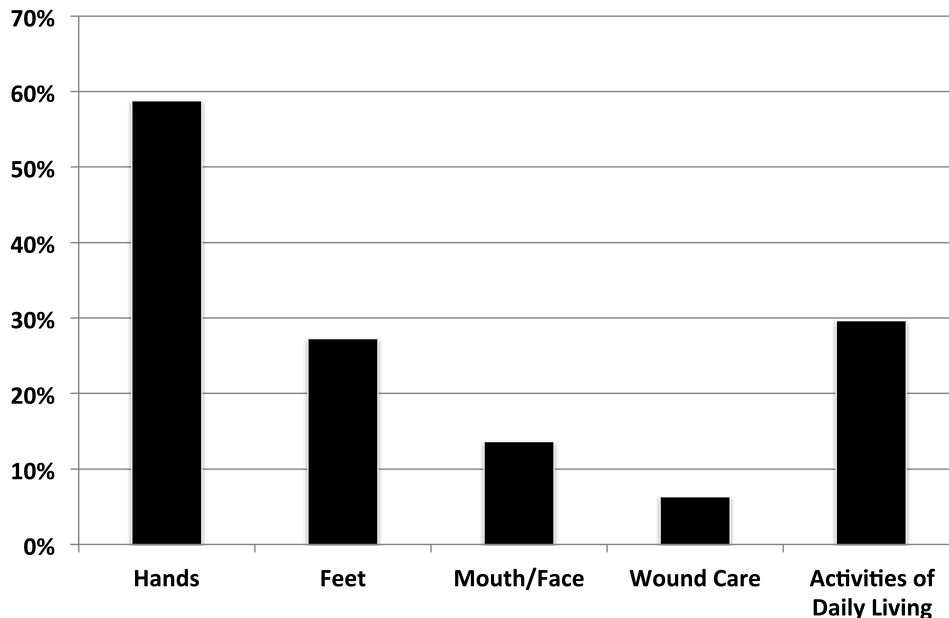


Figure 1. Physical and occupational therapy indications in the 3 months prior to enrollment in the Scleroderma Patient-centered Intervention Network (SPIN) cohort.

Table 1. Differences in demographic and medical variables between patients who used PT/OT and those who did not.

Variable	PT/OT, n = 381	No PT/OT, n = 1246	p
Age, yrs, mean (SD)	54.5 (12.7)	55.1 (12.4)	0.48
Sex			
Female	337 (24)	1086 (76)	Reference
Male	44 (22)	160 (78)	0.50
Country of enrollment			
USA	120 (17)	585 (83)	Reference
Canada	100 (23)	339 (77)	0.02
UK	32 (19)	137 (81)	0.56
France	119 (43)	159 (57)	< 0.01
Spain	10 (28)	26 (72)	0.10
Race/ethnicity*			
White	308 (23)	1034 (77)	Reference
Black	28 (24)	88 (76)	0.77
Other	45 (27)	123 (73)	0.27
Education, yrs, mean (SD)	15.1 (3.8)	15.0 (3.4)	0.78
Current occupation			
Employed	141 (20)	562 (80)	Reference
Unemployed	61 (28)	157 (72)	0.01
Retired	94 (24)	298 (76)	0.13
On disability	59 (36)	103 (64)	< 0.01
Other	26 (18)	121 (82)	0.51
Housing location			
Non-urban	225 (22)	784 (78)	Reference
Urban	155 (26)	444 (74)	0.10
Disease subtype			
Limited	179 (19)	750 (81)	Reference
Diffuse	188 (29)	453 (71)	< 0.01
Disease duration**, yrs, mean (SD)	10.4 (8.6)	11.7 (8.8)	0.02
Late disease, > 3 yrs	296 (23)	990 (77)	Reference
Early disease, ≤ 3 yrs	66 (29)	158 (71)	0.04
mRSS, mean (SD)	10.0 (9.9)	7.1 (7.8)	< 0.01
Raynaud phenomenon			
Negative	6 (25)	18 (75)	Reference
Positive	373 (23)	1218 (77)	0.86
Any digital ulcerations			
Negative	250 (24)	790 (76)	Reference
Positive	127 (23)	437 (77)	0.49
Tendon friction rubs			
Never	230 (21)	874 (79)	Reference
Currently	53 (31)	118 (69)	0.049
In the past	50 (27)	133 (73)	< 0.01
Small joint contractures			
No/mild, 0–25%	214 (19)	938 (81)	Reference
Moderate, 25–50%	103 (37)	176 (63)	< 0.01
Severe, > 50%	48 (42)	66 (58)	< 0.01
Large joint contractures			
No/mild, 0–25%	281 (21)	1043 (79)	Reference
Moderate, 25–50%	54 (40)	80 (60)	< 0.01
Severe, > 50%	21 (37)	36 (63)	0.01
ILD			
No	221 (22)	801 (78)	Reference
Yes	154 (27)	419 (73)	0.02
Pulmonary hypertension			
No	329 (24)	1026 (76)	Reference
Yes	39 (25)	119 (75)	0.91
Overlap syndromes			
Rheumatoid arthritis			
No	348 (23)	1149 (77)	Reference
Yes	30 (29)	72 (71)	0.16
SLE			
No	368 (24)	1185 (76)	Reference
Yes	12 (24)	39 (76)	0.98
Inflammatory myositis			
No	350 (23)	1157 (77)	Reference
Yes	25 (28)	64 (72)	0.29

Values presented as n (%) unless otherwise indicated. Response rate varied from 80% to 100% for the different variables. \*Consolidated race and ethnicity variable. \*\*Disease duration since first non-Raynaud manifestation. PT: physical therapy; OT: occupational therapy; mRSS: modified Rodnan skin score; ILD: interstitial lung disease; SLE: systemic lupus erythematosus.

Table 2. Differences in disability and psychological variables between patients who used PT/OT and those who did not.

Variable	PT/OT, n = 381	No PT/OT, n = 1246	p
Hand dysfunction (CHFS-II)	20.3 (18.9)	11.6 (14.6)	< 0.01
Disability index (SHAQ)	1.0 (0.7)	0.7 (0.7)	< 0.01
Depression (PHQ-8)	7.7 (5.8)	5.9 (5.3)	< 0.01
PROMIS-29			
Anxiety	54.1 (10.2)	51.3 (10.0)	< 0.01
Fatigue	57.5 (10.1)	54.4 (11.2)	< 0.01
Pain	59.3 (8.6)	54.5 (9.7)	< 0.01
Body image distress (SWAP)	35.6 (18.6)	30.1 (18.7)	< 0.01
Social anxiety (SIAS-6)	3.0 (4.6)	2.5 (3.8)	0.09
Self-efficacy (SEMCD)	5.8 (2.2)	6.7 (2.3)	< 0.01

Values presented as n (%) for categorical variables and mean (SD) for continuous variables. Response rate varied from 80% to 100% for the different variables. PT: physical therapy; OT: occupational therapy; CHFS-II: Cochin Hand Function Scale; SHAQ: Scleroderma Health Assessment Questionnaire; PHQ-8: Patient Health Questionnaire; PROMIS-29: Patient Reported Outcomes Measurement Information System; SWAP: Satisfaction with Appearance; SIAS-6: Social Interaction Anxiety Scale; SEMCD: Self-Efficacy for Managing Chronic Disease.

body image distress (SWAP  $35.6 \pm 18.6$  vs  $30.1 \pm 18.7$ ,  $p < 0.01$ ). Last, patients who used PT/OT had lower self-efficacy compared to those who did not (SEMCD  $5.8 \pm 2.2$  vs  $6.7 \pm 2.3$ ,  $p < 0.01$ ).

Table 3. Factors independently associated with PT/OT use in the SPIN cohort.

Variable	Model 1 <sup>a</sup>		Model 2 <sup>b</sup>	
	OR (95% CI)	p	OR (95% CI)	p
Age, yrs	1.00 (0.99–1.02)	0.54	–	–
Male sex	0.95 (0.62–1.46)	0.81	–	–
Country of enrollment				
USA			Reference	
Canada			1.62 (1.12–2.32)	0.01
UK			0.87 (0.52–1.45)	0.60
France			4.38 (2.99–6.42)	< 0.01
Spain	–	–	2.35 (0.99–5.58)	0.052
Education	1.04 (1.00–1.09)	0.04	1.08 (1.04–1.12)	< 0.01
Employment status				
Full time/part time			Reference	
Unemployed	1.11 (0.72–1.72)	0.63		
Retired	1.03 (0.68–1.57)	0.88		
On disability	1.09 (0.68–1.76)	0.71		
Other	0.68 (0.39–1.18)	0.17	–	–
Early SSc, disease duration $\leq 3$ yrs	1.24 (0.85–1.82)	0.26	1.43 (0.98–2.09)	0.06
Diffuse disease vs limited	1.12 (0.82–1.52)	0.48	–	–
Small joint contractures				
No/mild, 0–25%			Reference	
Moderate, 25–50%	2.11 (1.46–3.05)	< 0.01	2.09 (1.45–3.03)	< 0.01
Severe, > 50%	2.16 (1.28–3.64)	< 0.01	1.61 (0.95–2.72)	0.08
Large joint contractures				
No/mild, 0–25%			Reference	
Moderate, 25–50%	1.28 (0.80–2.05)	0.31	1.22 (0.76–1.96)	0.42
Severe, > 50%	1.77 (0.87–3.58)	0.11	2.33 (1.14–4.74)	0.02
Any digital ulcerations	0.68 (0.51–0.92)	0.01	0.70 (0.51–0.95)	0.02
Disability index (SHAQ)	1.23 (0.93–1.63)	0.15	1.54 (1.18–2.02)	< 0.01
Depression (PHQ-8)	1.02 (0.98–1.05)	0.34	–	–
Pain (PROMIS-29)	1.04 (1.02–1.06)	< 0.01	1.04 (1.02–1.06)	< 0.01
Self-efficacy (SEMCD)	1.00 (0.92–1.08)	0.93	–	–

<sup>a</sup>A priori defined model. <sup>b</sup>Reduced model following a stepwise regression procedure, adjusted for age and sex. PT: physical therapy; OT: occupational therapy; SSc: systemic sclerosis; SHAQ: Scleroderma Health Assessment Questionnaire; PHQ-8: Patient Health Questionnaire; PROMIS-29: Patient Reported Outcomes Measurement Information System; SEMCD: Self-Efficacy for Managing Chronic Disease.



The *a priori* defined and final multivariable regression models are described in Table 3. Using a backward stepwise technique, these factors were significantly associated with PT/OT use after adjusting for age and sex: higher education, more small joint contractures, fewer DU, higher disability, and more pain. There was an 8% increase in the likelihood of PT/OT use for every additional year of education (OR 1.08, 95% CI 1.04–1.12). The likelihood of PT/OT use was significantly higher in the presence of moderately severe small joint contractures (OR 2.09, 95% CI 1.45–3.03) and severe large joint contractures (OR 2.33, 95% CI 1.14–4.74). Higher disability (OR 1.54, 95% CI 1.18–2.02) and pain (OR 1.04, 95% CI 1.02–1.06) scores were associated with more PT/OT use. On the other hand, the presence of DU was found to decrease the odds of PT/OT use (OR 0.70, 95% CI 0.51–0.95). The association between PT/OT use and country of enrollment was analyzed after *a priori* variables were defined and included in the final model for analysis. Compared to patients from the United States, patients from Canada were 62% more likely to use PT/OT services (OR 1.62, 95% CI 1.12–2.32), while those from France were 4× more likely to use PT/OT (OR 4.38, 95% CI 2.99–6.42).

## DISCUSSION

In our present cross-sectional study, we found that 23% of patients with SSc in a large international cohort used PT/OT services in the 3 months prior to enrollment, and that hand PT/OT was the most common rehabilitation service used by these patients. Our findings also demonstrated that PT/OT use was significantly and independently associated with higher education, more severe musculoskeletal involvement, and higher disability and pain scores. We showed that geographical differences existed in the rate of PT/OT use.

Variable rates of PT/OT use in SSc have been described in the literature. Bassel, *et al* reported a rate of 28% PT/OT referral and 12% current use among 317 Canadian SSc patients with hand involvement surveyed between September 2008 and August 2009<sup>10</sup>. Consistent with our findings, the study by Bassel, *et al* also showed that the presence of more hand problems was associated with PT/OT referrals<sup>10</sup>. Through another survey study of 813 Canadian patients with SSc, Johnson, *et al* showed that 36% of patients have seen a physical therapist and 22% an occupational therapist since SSc diagnosis<sup>9</sup>. A higher rate of PT/OT use was shown in Western Europe. Among 198 Dutch patients with SSc surveyed from June and August of 2011, 75% reported contact with a physical therapist and 36% with an occupational therapist since SSc onset. Of these patients, 53% and 13% reported contact with a physical therapist and an occupational therapist, respectively, in the 12 months prior to study survey<sup>11</sup>. Between-study differences in sampling and methodology can explain some of the variability in the rates of PT/OT use. For example, in our study, the evaluation was restricted to the 3 months prior to enrollment while other

studies looked at use over longer periods of time. However, this variability is in accordance with our study findings of different rates of PT/OT use in the different countries evaluated, which reflects regional variations in SSc management, access to rehabilitation services, and healthcare costs.

Rehabilitation services use has been more extensively studied in RA, a more common rheumatic disease with a predominance of musculoskeletal manifestations. In RA, the rates of PT and OT use among 8001 German patients with RA were estimated at 44% and 15%, respectively, from 1993 to 1998<sup>26</sup>. Similar use rates were observed among 1200 patients with RA from Amsterdam in 1997 (40% PT and 17% OT use in the preceding year)<sup>27</sup>. In the United States, a 2011 study of 772 patients with RA showed that 15% of patients used PT in the preceding 6 months<sup>28</sup>. The variability can be explained by differences in the management and prognosis of RA between the pre- and post-biologics eras but might also reflect country differences in PT/OT use as seen in our study. Similar to our findings, disease activity, disability, and higher education were also predictive of PT use in RA<sup>28</sup>. In both RA and SSc, patients with these characteristics are in more need for (and thus more likely to be referred to) PT/OT and are possibly more likely to ask for or participate in these services because of their higher education and/or better access.

In the SPIN Cohort, hand PT/OT was the most commonly used rehabilitation service, reflecting the importance of hand symptoms in SSc. Hand pain and stiffness were previously shown to be frequently reported by patients with SSc, with more than 50% associating them with moderate to severe effect on quality of life<sup>3</sup>. In our study, patients who received PT/OT had more severe small joint contractures, tendon friction rubs, and higher hand dysfunction. Of interest, the presence of DU was independently associated with less PT/OT use. Wound care is recommended for the management of DU<sup>29</sup> and is offered by different specialists including specially trained physical therapists. However, wound care was an indication for PT/OT for about 6% of the SPIN cohort only, limiting conclusions on its association with DU. Based on limited studies, exercise had no direct effect on healing ulcers<sup>30</sup>. Because avoiding trauma is one of the recommended nonpharmacological interventions in the management of DU<sup>29</sup>, it is conceivable that concerns regarding exacerbation of ulcers by the trauma of PT/OT activities result in the hesitation of physicians to refer patients to PT/OT and of patients to participate in therapy when DU are active.

No association was seen between PT/OT use and age, sex, race/ethnicity, and overlap syndromes with other rheumatic diseases with significant musculoskeletal involvement. Although patients who used PT/OT were slightly more likely to have interstitial lung disease than not, no independent association was found between PT/OT use and SSc-associated cardiopulmonary disease.

Patients who used PT/OT were found to have higher psychological distress and lower self-efficacy than those who did not. Because all measures were obtained at the same timepoint, conclusions on a cause-effect relationship between distress, self-efficacy, and PT/OT use are not possible. The higher distress and lower self-efficacy may reflect a more severe disease and higher disease effect in those who were more likely to be referred to, and thus more likely to use, these services. In SSc, lower self-efficacy has been found to correlate with greater physical limitation, increased pain and fatigue, and more depression<sup>31</sup>. Moreover, better self-efficacy has been associated with better health outcomes, improved adherence to home exercise programs, and reduced health services use and cost in patients with chronic diseases including arthritis<sup>32,33,34</sup>. Most of these studies focused on hospitalizations and emergency room and physician visits.

The large sample size, number of variables, and *a priori* selected variables are important strengths. We included demographic variables, disease-specific variables including a detailed assessment of musculoskeletal manifestations, disability measures, and a number of psychological variables that were thought and/or previously shown to affect health services use. SSc diagnosis and medical variables were ascertained by physicians and all variables were provided at 1 timepoint, allowing for a real-time assessment of their association with PT/OT use.

There are important limitations to consider when interpreting the findings of our study. The SPIN Cohort is a convenience sample of patients recruited from specialized SSc centers, which might limit the generalizability of our findings. These patients have access to tertiary healthcare systems and are likely different from those seen in other settings. The rate of PT/OT use might therefore be lower in patients receiving care in generalized rheumatology practices. As previously stated, we were able to look at PT/OT use only in the 3 months prior to enrollment, and the evaluation of PT/OT referrals was limited. A previous study showed differences between the rates of PT/OT referrals and use, indicating possible differences in the perceived need for PT/OT between physicians and patients, among other factors worth investigating. In SPIN, questions on PT/OT use were restricted to supervised rehabilitation services only and might not have identified home-based physical therapy and exercise, which have been reported by more than half of patients with SSc in the SPIN cohort in a study by Azar, *et al*<sup>35</sup>. In addition, it is important to recognize that the lack of data on income, medical insurance, and access to rehabilitation services is an important limitation of our study, given the critical effect these variables have been shown to have on PT/OT and other health services use. In adult patients with low back pain in the United States, out-of-pocket expenditure and type of insurance were shown to be predictive of PT visits, with privately or Medicare-insured people more likely to have more visits compared to those publicly insured or

uninsured<sup>36</sup>. Similarly, SSc patients with private insurance were almost twice as likely to be referred to PT/OT in the study by Bassel, *et al*<sup>10</sup>. Because of the complexity and variability of insurance coverage within each healthcare system, looking at the rate of PT/OT use across the different countries does not fully overcome this limitation.

We showed that fewer than 25% of patients with SSc in a large international cohort used PT/OT services in the 3 months prior to enrollment and that geographical differences exist in the rate of use. Future research evaluating the effect of rehabilitation services on functional outcomes in SSc is needed to fully understand the importance of these interventions in SSc and to strengthen the evidence base for development of management guidelines. Further, in addition to identifying barriers to PT/OT in different healthcare settings, interventions are needed to improve the use of these services among patients with SSc. Such interventions include developing effective SSc-specific online and home-based physical therapy and exercise programs, which is one of the primary aims of SPIN. These programs could help overcome some of the barriers to PT/OT use, including cost and access.

## APPENDIX 1.

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